Rare conditions mimicking bladder endometriosis

Devi Balasubramaniam[1], Kavitha Yogini Duraisamy[2], Malathi Ezhilmani[3], Aravinth Subramanium[4]

From [1]Consultant, [2]Head, [3]Fellow, Department of Endogynecology, [4]Head, Department of Pathology, GEM Hospitals and Research Centre, Coimbatore, Tamil Nadu, India

Correspondence to: Dr. Devi. B, GEM Hospital and Research Center, 45A, Pankaja Mills Road, Ramanathapuram, Coimbatore, Tamil Nadu – 641045, India. E-mail: devibalasubramaniam@gmail.com

Received - 06 October 2018 Initial Review - 24 October 2018 Accepted - 16 November 2018

ABSTRACT

Benign conditions of the bladder such as cystitis cystica and Xanthogranulomatous cystitis (XC) are chronic inflammatory lesions which are usually rare in incidence. Here, we report 2 case reports wherein both the patients were diagnosed as bladder endometriosis for which segmental resection and partial cystectomy were done by the minimally invasive approach. Post-operative pathological examination revealed the lesions as cystitis cystica in one case and XC in another. To the best of our knowledge, this case of cystitis cystica mimicking as the bladder endometriosis is the 2nd case reported in the literature and case of XC forming a tumorous lesion is the 29th to be reported in the world literature.

Key words: *Cystitis cystica, Xanthogranulomatous cystitis, Bladder endometriosis, Robotic, Laparoscopic partial cystectomy*

Xanthogranulomatous cystitis (XC) is a rare chronic granulomatous inflammation of the bladder [1]. It is characterized by lipid-laden macrophages known as xanthoma cells, multinucleated giant cells, cholesterol clefts, fibrosis, and calcification. Similarly, another unusual entity, cystitis cystica is a benign chronic inflammatory condition which is characterized by the proliferative changes in the urothelium on long-term irritation leading to multiple small filling defects in the bladder wall. It is most commonly found in the trigone area. In general, it causes no symptoms while symptoms of urinary infection can occur. It is not a rare disease, but cases forming a tumorous lesion with blueberry spots suspected to be caused by endometriosis were reported previously in just one case report in the literature.

We, thus present two case reports of unusual conditions mimicking bladder endometriosis. To the best of our knowledge, this case of cystitis cystica mimicking as the bladder endometriosis is the 2nd case reported in the literature and case of XC forming a tumorous lesion is the 29th to be reported in the world literature [2].

CASE REPORT

Case 1

A 32-year-old case of primary infertility, with endometriosis since 4 years, was referred for management of bladder endometrioma which was diagnosed 2 weeks back by imaging. She had regular cycles, with mild tolerable pain. She had a history of fever for 1 week followed by severe pain at the end of micturition for 1 month following ovum pick up. The patient had a history of pyuria lasting for 1 week which was treated with antibiotics.

The patient was stable, pulse 88, blood pressure of 120/70 mm Hg and respiratory rate 16. Blood investigations were within normal limits. On evaluation, magnetic resonance imaging (MRI) suggested thick-walled cystic lesion 10 cm ×6 cm × 5.5 cm superior to the urinary bladder with disruption of the right lateral wall of the cyst. Along with that, extra-cystic fluid loculations adherent to the bladder wall, with no obvious fistulous communication suggestive of a chocolate cyst extending to the urinary bladder.

Intraoperatively, cystoscopy confirmed the infiltration involving mucosa and adventitial layer, seen well away from the interureteric ridge. Large left ovarian endometriotic cyst of 10 cm × 8 cm densely adherent to the dome of the bladder (Fig. 1) was noted by laparoscopy. After adhesiolysis and ovarian cystectomy, nodular bluish lesion of 3 cm ×3 cm was noted over bladder dome, which appears infiltrating the bladder wall (Fig. 2). In view of prolonged infertility, deep infiltrating endometriosis and an inability to provide with long term hormonal treatment, she was taken up for laparoscopic segmental resection of the bladder endometrioma. Histopathology revealed XC showing foamy macrophages with no evidence of endometrial glands or stroma (Fig. 3). Foleys catheter was removed after 2 weeks. The patient is on regular follow-up and symptom-free till date and referred to the infertility center for further management.

Case 2

A 34-year-old multipara, known epileptic, had regular cycles without dysmenorrhea presented with painful micturition, increased frequency, and urgency during cycles which persist till day 10 of the cycle for the past 5 years. For that, she was evaluated 2 years back elsewhere and was confirmed as bladder

endometriosis following cystoscopy guided biopsy. Since then, she was on dienogest and was symptom-free.

She developed urinary frequency with strangury and dysuria for past 1-month in spite of hormonal induced amenorrhea which was not responding to antibiotics, and urine culture was sterile. Her pelvic examination revealed shallow anterior fornix with induration. A vaginal scan showed diffuse thickening of the posterior wall of bladder and MRI pelvis showed 4 cm bladder wall endometrioma with the transmural involvement of posterior wall of the bladder without infiltration of the anterior uterine myometrium. As the patient had a relapse of symptoms in spite of hormonal therapy and also she was reluctant to continue long-term hormones, we decided to prepare her for a robotic approach in view of more precise excision and suturing. She underwent cystoscopy guided robotic partial cystectomy (Fig. 4).

Intraoperative cystoscopy revealed normal external meatus, urethra, and bilateral ureteric orifices. Around 5 cm × 2.5 cm bluish nodular endometriotic lesion was noted in the posterosuperior wall of the bladder involving the muscularis layer. A bulge was seen tenting toward the lumen of the bladder, inferior margin of the lesion was 4 cm away from interureteric ridge (Fig. 5). Intraoperatively, no evidence of pelvic endometriosis was observed, and the lesion from the bladder was not found to involve the uterine wall. Bilateral ureteric stenting was done before surgery; the lesion was completely excised with a 0.5 cm wide

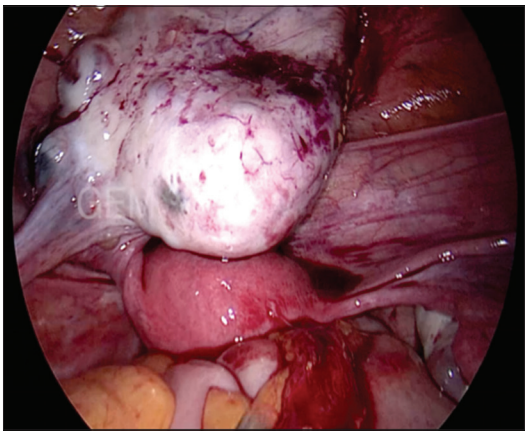

Figure 1: Ovarian endometriotic cyst adherent to the bladder surface

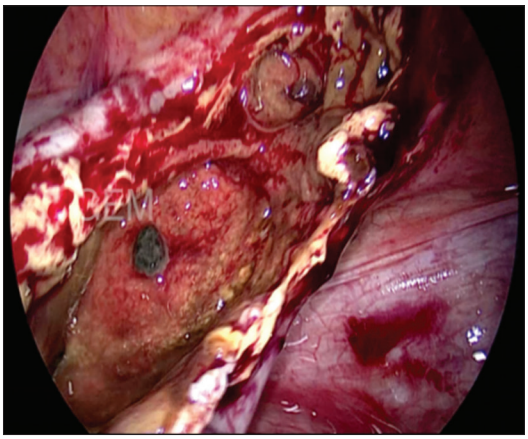

Figure 2: Urinary bladder lesions following ovarian cystectomy

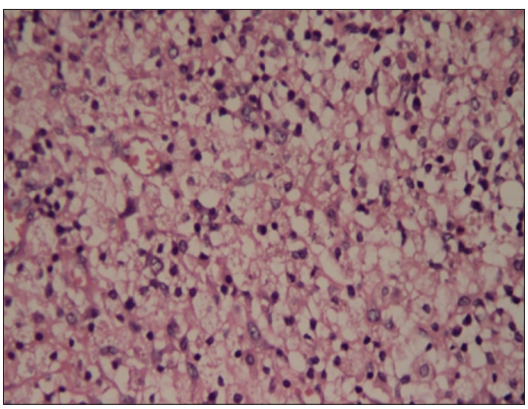

Figure 3: High power field showing foamy macrophages

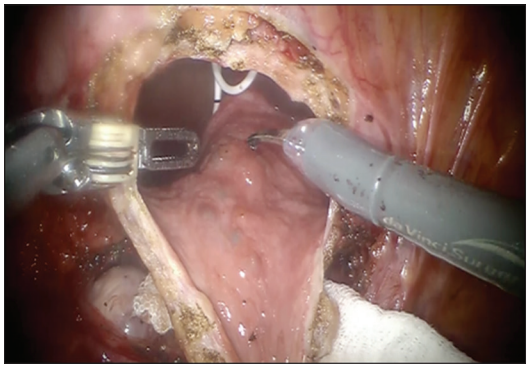

Figure 4: Excision of the entire lesion through Robotic partial cystectomy

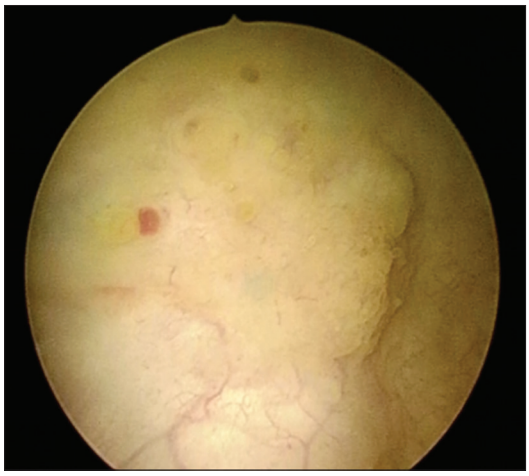

Figure 5: Cystoscopic view of the bladder lesion

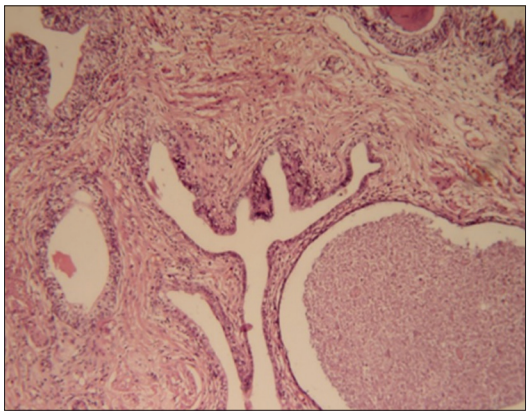

Figure 6: High power view of cystitis cystica

margin of healthy tissues. Bladder closure was done in 2 layers using 3–0 Vicryl and ureteric stents were removed. Histopathology of the specimen suggested cystitis cystica (Fig. 6). Foleys catheter was placed *in situ* for 3 weeks, and cystoscopy was repeated after 3 weeks. There were no abnormal findings, and the patient is on follow-up and now symptom-free without any hormones till date.

DISCUSSION

XC is a rare benign inflammatory disease [1]. Histologically, Xanthogranulomatous lesions can be misinterpreted as malakoplakia. The symptoms of XC are nonspecific and difficult to differentiate from other pathologies of the bladder, especially malignancy. Chronic irritation caused by endometriosis may be the reason as in our first case, the patient presented with ovarian endometrioma. It often presents with symptoms of cystitis and may be associated with carcinoma [3]. Hence, it is important that the surgeon should be aware of this unusual dual pathology. Furthermore, it is essential that the pathologists should study entire specimen during gross examination. The curative treatment of choice is surgery [4]. Partial cystectomy is preferred and is the most commonly performed procedure. Endoscopic excision has also been reported for small lesions [5,6].

Cystitis cystica is a benign proliferative lesion of the bladder; caused as a result of a chronic reactive inflammatory disorder due to irritation of the urothelium. It can occur at any age and characterized by hyperplasia of the bladder mucosa and presence of cystic dilation of Von Brunn nests within the lamina propria. Minor forms are common and have similar features as cystitis, but its major form may be mistaken for bladder tumor on cystoscopy. The different types are cystitis cystica, cystitis glandularis of a common type and cystitis glandularis of the intestinal type [7].

Cystitis glandularis of the intestinal type is usually misdiagnosed as adenocarcinoma, but there is no significant atypia, no muscular invasion, and no/minimal mitotic activity [8]. It occurs in trigone, ureter, and renal pelvis. Usually asymptomatic, may cause recurrent urinary tract infections; often benign incidental findings in biopsies done for other reasons. Although the etiology remains unknown, it has been postulated to be chronic stimulation of the bladder mucosa and glandular metaplasia of transitional epithelial cells [9].

Chronic urinary tract infections, inflammation caused by urolithiasis, tumor, outflow obstruction, and indwelling urethral or suprapubic catheter are the risk factors which were not present in our case. It is not a rare disease, but a case forming a tumorous lesion with blueberry spots suspected to be endometriosis was a rare appearance of the disease, reported once in the past literature. Chronic inflammation of the bladder by endometriosis of the uterus may result in proliferative change forming an extravesical mass with wall penetration. Moreover, frequent submucosal bleedings occurred by chronic inflammation of the bladder could cause the formation of blueberry spots. As the disease originates outside the bladder according to Vercellini *et al*., 2002, transurethral resection will result in incomplete excision, because radicality would imply bladder perforation [10].

Cystitis cystica has been regarded as a potentially premalignant condition predisposing to adenocarcinoma of the bladder [11]. In contrast, it was reportedly found in 93% of normal autopsied patients. These findings have suggested that it is a normal variant of bladder epithelium rather than a premalignant lesion. However, these autopsy series had no gross findings and were only microscopic findings. It has been reported that clinically evident disease with macroscopic findings could be a progression of the disease and premalignant lesions [9]. Chapron *et al*. stated that there are no indications for TUR in case of deep infiltrating bladder endometriosis. It is, therefore, better to avoid TUR in patients presenting with deep infiltrating bladder endometriosis [12].

Only a transurethral resection (TUR) is performed in most cases for diagnosis and treatment of cystitis cystica. However, there have been some cases in which total cystectomy were performed due to recurrence or continuance of tumor and symptoms despite frequent TUR of the tumor [13].

Medical management alone in the treatment of deep infiltrating bladder endometriosis will result in the recurrence rate of 70% [14]. Seracchioli *et al*. found that TUR of deep infiltrating bladder endometriosis results in incomplete resection, which eventually result in recurrence. Partial cystectomy/ segmental resection involves removing the entire vesical lesion and prevent recurrence. Sener *et al*. stated that generally deep infiltrating bladder endometriosis will be associated with posterior and anterior pouch endometriosis such as uterosacral and rectovaginal septum, which will be treated concurrently if we combine laparoscopy and cystoscopy approach [15] For deep infiltrating bladder endometriosis we believe, and as per many authors' statement, the best modality of treatment is laparoscopy/ abdominal approach and complete excision of the lesion.

CONCLUSION

Chronic benign lesions such as XC and cystitis cystica can present with the bladder mass and symptoms similar to the bladder endometriosis. Hence, it is important that the surgeon should be aware of this unusual dual pathology and should keep in mind about the malignant potential of these chronic lesions of the urinary bladder.

REFERENCES

1. Goel R, Kadam G, Devra A. Xanthogranulomatous cystitis. Int Urol Nephrol 2007;39:477-8.
2. Pengliang C, Fengsheng L, Siyao H, Wenjie L, Fei L. A case report of xanthogranulomatous cystitis. Urol Case Rep 2017;13:72-3.
3. Bates AW, Fegan AW, Baithun SI. Xanthogranulomatous cystitis associated with malignant neoplasms of the bladder. Histopathology 1998;33:212-5.
4. Hayashi N, Wada T, Kiyota H, Ueda M, Oishi Y. Xanthogranulomatous cystitis. Int J Urol 2003;10:498-500.
5. Yang KS, Kim YH, Seong YK, Kim IG, Han BH, Kim SJ. Xanthogranulomatous cystitis arising from the posterior wall of the bladder. Korean J Urol 2011;52:868-9.
6. Singh SK, Khandelwal AK, Pawar DS, Sen R, Sharma S. Xanthogranulomatous cystitis: A rare clinical entity. Urol Ann 2010;2:125-6.
7. Rau AR, Kini H, Pai RR. Morphological evaluation of cystitis glandularis.

Indian J Pathol Microbiol 2009;52:203-5.
8. Young RH, Bostwick DG. Florid cystitis glandularis of intestinal type with mucin extravasation: A mimic of adenocarcinoma. Am J Surg Pathol 1996;20:1462-8.
9. Semins MJ, Schoenberg MP. A case of florid cystitis glandularis. Nat Clin Pract Urol 2007;4:341-5.
10. Vercellini P, Frontino G, Pisacreta A, De Giorgi O, Cattaneo M, Crosignani PG. The pathogenesis of bladder detrusor endometriosis. Am J Obstet Gynecol 2002;187:538-42.
11. Edwards PD, Hurm RA, Jaeschke WH. Conversion of cystitis glandularis to adenocarcinoma. J Urol 1980;108:568-70.
12. Chapron C, Bourret A, Chopin N, Dousset B, Leconte M, Amsellem-Ouazana D, *et al*. Surgery for bladder endometriosis: Long-term results and concomitant management of associated posterior deep lesions. Hum Reprod 2010;25:884-9.
13. Goff WB. Cystitis cystic and cystitis glangularis: Cause of bladder mass. J Comput Assist Tomogr 1983;7:347-9.
14. Nezhat CH, Nezhat FR, Nezhat CR, Malik S, Osias J. Laparoscopic management of 15 patients with infiltrative endometriosis of the bladder and a case of primary intravesical adenosarcoma. Fertil Steril 2002;78:872-5.
15. Sener A, Chew BH, Duvdevani M, Brock GB, Vilos GA, Pautler SE. Combined transurethral and laparoscopic partial cystectomy and robot-assisted bladder repair for the treatment of bladder endometrioma. J Minim Invasive Gynecol 2006;13:245-8.

Funding: None; Conflict of Interest: None Stated.

How to cite this article: Balasubramaniam D, Duraisamy KY, Ezhilmani M, Subramanium A. Rare Conditions Mimicking Bladder Endometriosis. Indian J Case Reports. 2018;4(6):450-453.

Doi: 10.32677/IJCR.2018.v04.i06.013